Title
Unsuspected tarsal coalitions in congenital clubfoot and equinus

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ABSTRACT:

Purpose: Tarsal coalitions have been reported to occur in the setting of clubfeet, but only as rare isolated findings. Other varus foot deformities have a known association with tarsal coalition, such as calcaneonavicular coalitions with spastic pes varus. The purpose of this paper was to report a two-institution count on the number of tarsal coalitions seen in congenital and neurogenic clubfeet, to report the break-down of types of coalitions encountered, and to suggest methodology to improve earlier diagnoses.

Methods: The records of all patients treated by two of the authors for bilateral clubfoot or equinus and found to have a tarsal coalition between 2006 and 2016 were reviewed. An incidence rate was calculated using the number of patients treated for congenital clubfoot or equinus without tarsal coalition during the same time period.

Results: Thirteen feet with tarsal coalition (calcaneonavicular n=11 and talocalcaneal n=2) were reviewed. The incidence rate of tarsal coalition in the setting of congenital clubfoot or equinus at the two institutions was 2.6 and 4.0%. 38% of coalitions were found in otherwise healthy children with bilateral congenital clubfoot, 31% in children with neurogenic clubfoot due to underlying spasticity, and 31% in children with pure equinus deformity. 77% of cases were calcaneonavicular coalitions, and the remaining 23% talocalcaneal; there was no significant difference in coalition type between groups. All patients were definitively diagnosed by CT scans with 3D reconstruction, often after physical exam and/or oblique radiographs increased suspicion. In 38% of cases, the patient had previously undergone at least one open procedure before the coalition was recognized. The mean age at diagnosis was 11.4 years.

Conclusions: Our experience suggests that tarsal coalitions, particularly calcaneonavicular coalitions, may occur more frequently in clubfoot and equinus deformities than previously reported. As tarsal coalitions are easy to miss on physical examination of a patient with rigid foot deformity, frequent use of oblique radiographs and CT scans as necessary should result in earlier detection of tarsal coalitions and better foot correction.

Level of Evidence: IV, Retrospective Review.

Keywords clubfoot, equinus, rigid foot deformities, talipes equinovarus, tarsal coalitions.

FULL TEXT

BACKGROUND:

Tarsal coalition has previously been reported in patients with congenital clubfoot (1,2,3). Although rare, it adds an additional degree of complication in the treatment of rigid equinovarus foot deformities and thus is an important anatomic abnormality to identify before operative interventions. In scattered reports of tarsal coalitions discovered in congenital clubfoot, the majority have been talocalcaneal (TC) coalitions with occasional naviculocuneiform coalitions (1,2,4). There are almost no reports of calcaneonavicular (CN) coalitions, which is surprising given that CN coalitions are the most common type of tarsal coalition in the general population (5).

The first report of tarsal coalition in the setting of clubfoot deformity was described by Robert Callahan in 1980. Prior to his report, tarsal coalitions had been described in association with other varus foot deformities, particularly pes varus (6,7), and it had been hypothesized that tarsal coalitions can lead to a varus deformity via spasm of the tibialis muscles (6). Callahan described a 14 month-old boy with congenital clubfoot, a talocalcaneal coalition, and absent posterior tibialis tendon (3), suggesting that an equinovarus deformity could arise from other pathologic mechanisms and that these mechanisms might be independently associated with tarsal coalitions. To date, these pathologic mechanisms are not well understood and are likely multifactorial.

There has been one case series since Callahan's original report, in which Spero and Tornetta described 17 cases of TC coalition and a single case of CN coalition in patients

with congenital clubfoot (1). Experience at our institution differs from this report because Spero and Tornetta described mostly TC coalitions, diagnosed at an average age of 17 months, whereas the senior authors were regularly diagnosing CN coalitions in significantly older patients (7-15 years) with congenital clubfeet or equinus. Given our contrasting experience, the purpose of this paper is to provide a two-institution incidence rate on the number of tarsal coalitions in congenital clubfoot or equinus, to report the break-down of coalition types encountered, and to suggest methodology for earlier diagnosis. In doing so, we aim to increase awareness about tarsal coalitions in the setting of rigid pediatric foot deformities such that patients may benefit from earlier diagnosis and interventions as needed.

METHODS:

The records of nine patients with bilateral clubfoot or pure equinus deformity and a diagnosis of unilateral or bilateral tarsal coalitions were reviewed. All cases were treated by two of the authors. All clinic notes, operative reports, and radiographs were retrospectively reviewed. Data collected included quality and laterality of congenital foot deformity, quality and laterality of tarsal coalition, reported symptoms and level of functioning, when and how the coalition was initially diagnosed (i.e. routine radiographs, physical exam findings which prompted additional radiographs, discovery in the operating room, etc.), interventions prior to diagnosis, and interventions since diagnosis. Medical conditions affecting the musculoskeletal and/or nervous systems were also recorded.

RESULTS:

Thirteen feet with tarsal coalition were found in nine patients with foot deformities of talipes equinovarus or equinus. Eleven of these cases came from an institution in which 273 patients were treated for congenital or neurogenic clubfoot or equinus over the same time period, making an incidence rate of approximately 4.0%. Two of the cases came from an institution in which 78 patients were treated for the same conditions, making an incidence rate of approximately 2.6%. Five of the nine patients were found to have unilateral tarsal coalitions and four patients had bilateral coalitions. All were definitively diagnosed by computed tomography at an average age of 11.4 years. One patient had suspicious findings on physical exam that prompted further radiographic evaluation, while the remainder were incidental findings on X-ray (lateral ankle for TC coalitions or oblique foot for CN coalitions) or pre-operative planning CT. All patients had stiffness/pain and/or refractory pathology such as equinus or equinovarus. In five cases (38% of feet), the patient had previously undergone at least one open procedure before the coalition was recognized.

Tarsal coalitions were found in patients with rigid foot deformities in one of three categories: congenital clubfoot (Table 1), neurogenic clubfoot (Table 2), or isolated equinus deformity (Table 3). In the congenital clubfoot group, four otherwise healthy children were born with varying degrees of talipes equinovarus requiring at least serial manipulations and casting as an infant and sometimes open procedures as well. In the neurogenic clubfoot group, three patients presented with varying degrees of equinocavovarus. In the equinus group, two patients presented with extreme toe walking

and straightforward bilateral equinus deformity secondary to an Achilles contracture and no associated cavus, varus or adductus. None of the tarsal coalitions were iatrogenic from the prior treatment.

1. Congenital Clubfoot

Thirty-eight percent of the tarsal coalitions (five feet) were found in four patients with congenital clubfeet treated with serial manipulations and casting. Two out of the five feet were also treated with a posteromedial release at ages 5 months and one year respectively. The coalitions were not recognized until ages 10-12 years (mean = 11.2 years). At the time of coalition recognition, all five patients had pronounced equinocavovarus deformities (Fig. 1, 2).

Patient 1 was a 10 year-old male with no prior foot surgeries and bilateral clubfeet consisting of moderate equinovarus bilaterally. While planning surgical correction, lateral X-rays and 3D CT revealed bilateral CN coalitions (Fig. 1). These were both resected prior to reconstructive foot surgery.

Patient 2 was a 12 year-old male with bilateral congenital clubfeet consisting of moderate equinovarus, who had previously undergone posteromedial release at age 5 months. He complained of persistent right anterolateral foot pain and was noted to have decreased subtalar motion on the right compared to the left. Oblique X-rays revealed a unilateral CN coalition, which was confirmed by CT scan and subsequently resected.

Patient 3 was a 12 year-old male with bilateral clubfeet consisting of pronounced equinovarus, who had previously undergone posteromedial release at age one year. Weight-bearing oblique X-rays revealed a unilateral CN coalition which was previously missed on non-weight bearing AP and lateral films. The coalition was confirmed by CT scan and subsequently resected.

Patient 4 was a 12 year-old male with no prior foot surgeries and bilateral clubfeet. At presentation, his right foot demonstrated a significant cavovarus deformity. Suspicious radiographs led to CT scans and the diagnosis of a unilateral TC coalition (Fig. 2). This was subsequently resected.

2. Neurogenic Clubfoot

Thirty-one percent of the coalitions (four feet) were found in three patients with clubfoot due to a neurologic condition (Fig. 3).

Patient 5 was a 12 year-old female with scoliosis, developmental delay, spasticity, and bilateral clubfoot. She had previously undergone casting in infancy and multiple surgical interventions including bilateral posteromedial release at 6 months, additional bilateral surgical correction at 2 years and right foot osteotomies at 11.5 years. After this, she continued to have some residual cavovarus deformity. At a routine follow-up appointment following the foot osteotomies, an oblique right foot x-ray revealed a unilateral calcaneonavicular coalition. After confirmatory CT scans, it was resected.

Patient 6 was a 12 year-old male with Charcot Marie Tooth and bilateral equinovarus deformity, who had previously undergone bilateral Achilles tendon lengthening at age 9 years. While planning surgical correction for his feet, a pre-op CT demonstrated CN coalitions bilaterally (Fig. 3). The more severe of the coalitions was surgically removed before his first reconstructive foot surgery.

Patient 7 was an 11 year-old female with no significant past medical history who presented with chronic left foot and heel pain and mild cavovarus deformity. X-rays revealed a likely talocalcaneal coalition, which was confirmed by CT scan and subsequently resected. One year after resection, her cavovarus deformity recurred and she was noted to have continued muscle weakness despite physical therapy. At that time, it was discovered that the patient's father had been diagnosed with a neuropathy in his early 20s, most likely Charcot Marie Tooth. The patient was referred to neurology to confirm that diagnosis.

3. Equinus only

Thirty-one percent of the coalitions were found in two patients with only severe equinus (Fig 4). Both had remarkable toe walking and no prior surgery.

Patient 8 was a 7 year-old female with bilateral equinus and no other abnormalities. Her equinus measured near 20 degrees bilaterally. Her exam also revealed decreased subtalar motion and a palpable ridge over her sinus tarsi bilaterally. Lateral and oblique foot X-rays and CT scans confirmed her bilateral CN coalitions (Fig. 4). She subsequently

underwent CN coalition resection and Vulpius tendo-achilles lengthening on her left, followed by the same procedure on her right three months later.

Patient 9 was a 15 year-old male with autism, ADHD and severe bilateral equinus of 30 degrees. Pre-operative CT scans revealed bilateral CN coalitions. Surgical correction is being planned.

DISCUSSION:

In this paper, we have presented a series of thirteen feet with tarsal coalitions in nine patients with equinovarus or equinus deformity. The incidence rate for tarsal coalitions in the setting of these deformities was found to be 2.6% and 4.0% at our two institutions. Our study includes patients with tarsal coalitions in congenital clubfoot, neurogenic clubfoot, and pure equinus. This suggests that the coalitions are not the cause of the deformity, but an independent variable occasionally associated with it. All were typical coalitions with no suggestions of being iatrogenic in origin. While a few TC coalitions in the setting of congenital clubfoot have previously been described in the literature, reports of CN coalitions are extremely rare. This contrasts with findings in this series, in which the majority of the coalitions cases were CN (n=11). This suggests that CN coalitions may be more frequent than previously reported.

This study highlights the difficulty of diagnosing tarsal coalitions in the setting of rigid equinovarus and equinus due to non-specificity of symptoms, globally reduced motion on physical exam, and the need for appropriate radiographs (to include oblique images) in

order to visualize the coalition. Due to rarity of this problem, appropriate radiographs were not obtained or the findings missed until an average age of 11.4 years, when they were definitively diagnosed by senior orthopedic surgeons aided by 3D CT reconstruction Although one patient presented with persistent unilateral foot pain, foot pain is very commonly associated with the residuals of talipes equinovarus and thus unlikely to trigger an aggressive work up. An experienced orthopedic surgeon can recognize decreased subtalar motion, but in a paralytic and/or prior-operated, stiff equinovarus foot, reduced subtalar motion is very difficult to appreciate. In addition, if the patient has a bilateral coalition, their range of motion may be symmetrically reduced, further decreasing the likelihood of detection. In five cases, patients had previously undergone at least one open procedure before the coalition was recognized. Many had prior X-rays which failed to demonstrate the coalition. In the infant, it is unlikely that the coalition would even show, as ossification of the bony bridge does not occur until near age ten. After diagnosis and resection of the coalitions, the authors felt that surgical correction of the equinovarus was much more complete.

In summary, this study suggests that tarsal coalitions in the setting of equinus or equinovarus foot deformity may be more frequent than previously thought. The authors anticipate that earlier detection and resection of tarsal coalitions will enable better foot correction, increased range of motion and fewer operations. Frequent use of oblique radiographs and CT scans as necessary should result in additional cases.

Limitations of this study include the typical limitations of retrospective study. The patients were already diagnosed with one or more coalitions by the senior authors and thus, due to missed diagnoses, the actual incidence rate may be higher than the 2.6-4.0% that we have reported here. Further research is needed to explore the prevalence of various tarsal coalitions in talipes equinovarus and equinus.

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FIGURE LEGENDS:

Figure 1: Patient 1: CN coalition in a 10 year-old male with congenital clubfoot and no prior foot surgeries. Fig 1A-Lateral view of the right ankle demonstrates lengthening of the calcaneal anterior process and irregularities of the calcaneal and navicular borders, suggestive of CN coalition. Fig 1B- CT scan with 3D reconstruction confirms the presence of a classic CN coalition.

Figure 2: Patient 4: TC coalition in a 12 year-old male with congenital clubfoot and no prior foot surgeries. Fig 2A-Lateral view of the right foot demonstrating the "C sign," suggestive of TC coalition. Fig 2B- CT scan with 3D reconstruction confirmed the presence of TC coalition.

Figure 3: Patient 6: CN coalitions in a 12 year-old male with Charcot Marie Tooth and refractory equinovarus deformities after bilateral Achilles tendon lengthening. CT scans with 3D reconstruction (ordered for pre-operative planning) revealed bilateral CN coalitions (Left foot shown).

Figure 4: Patient 8: CN coalition in a 7 year-old female with congenital equinus and no prior foot surgeries. Fig 4A,B-Oblique and standing lateral views of the left foot demonstrate elongation of the anterior process

of the calcaneus, suggestive of a CN coalition. Fig 4C-CT scan with 3D reconstruction confirms the presence of CN coalition.

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Table 1. Congenital clubfoot

Patient	**Gender**	**PMH**	**Deformity**	**Tarsal Coalition**	**Treatment prior to coalition diagnosis**	**Age at coalition diagnosis (yrs)**	**Treatments since coalition diagnosis**
Pt. 1A	Male		Congenital clubfoot	Left Calc-Navic	Infancy: Serial casting	10	-15.6 yrs: Triple C -15.7 yrs: HWR
Pt. 1B				Right Calc-Navic	Infancy: Serial casting	10	-15.6 yrs: Triple C -15.7 yrs: HWR, I&D
Pt. 2	Male		Congenital clubfoot	Right Calc-Navic	-Infancy: Serial casting -5 months: Posteromedial release	12.15	None
Pt. 3	Male		Congenital clubfoot	Left Calc-Navic	-Infancy: Serial casting -1 yr: Bilateral posteromedial release	11	
Pt. 4	Male		Congenital clubfoot	Right Talo-Calc	Infancy: Serial casting	12	
Key: Triple C = Cuboid, cuneiform and calcaneal pin osteotomies; HWR = Hardware removal, I&D = Incision and drainage for surgical site infection.							

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Table 2. Neurogenic clubfoot

Patient	**Gender**	**PMH**	**Deformity**	**Tarsal Coalition**	**Treatment prior to coalition diagnosis**	**Age at coalition diagnosis (yrs)**	**Treatments since coalition diagnosis**
Pt. 5	Female	-Idiopathic scoliosis -Mild developmental delay	Neurogenic clubfoot (spastic paralytic)	Right Calc-Navic	-Infancy: Serial casting -6 months: Bilateral clubfoot correction -2 yrs: Additional bilateral clubfoot correction -13 yrs: Triple C	12.0	None
Pt. 6A	Male	Charcot Marie Tooth	Neurogenic equinovarus	Left Calc-Navic	9 yrs: Bilateral TAL	11.9	12.0 yrs (concurrent with coalition resection): TAL, FHL and FDL lengthening, PMR, lengthening of aberrant soleus muscle
Pt. 6B				Right Calc-Navic	9 yrs: Bilateral TAL	11.9	12.1 yrs: TAL, PMR, PT and FDL lengthening
Pt. 7	Female	Possible Charcot Marie Tooth	Neurogenic cavovarus	Left Talo-Calc	None	11.3	None

Key: Triple C = cuboid, cuneiform and calcaneal pin osteotomies; TAL = tendo-achilles lengthening; FHL = Flexor Hallucis Longus; FDL = Flexor Digitorum Longus; PT = Posterior Tibialis; PMR = posteromedial release

Table 3. Pure equinus

Patient	Gender	PMH	Deformity	Tarsal Coalition	Treatment prior to discovery	Age at discovery (yrs)	Treatments since discovery
Pt. 8A	Female		Equinus Only	Left Calc-Navic		7.4	Infancy: Serial casting
Pt. 8B				Right Calc-Navic		7.4	Infancy: Serial casting
Pt. 9A	Male	Autism spectrum disorder	Equinus Only	Left Calc-Navic		15.0	
Pt. 9B				Right Calc-Navic		15.0	

FIGURES:

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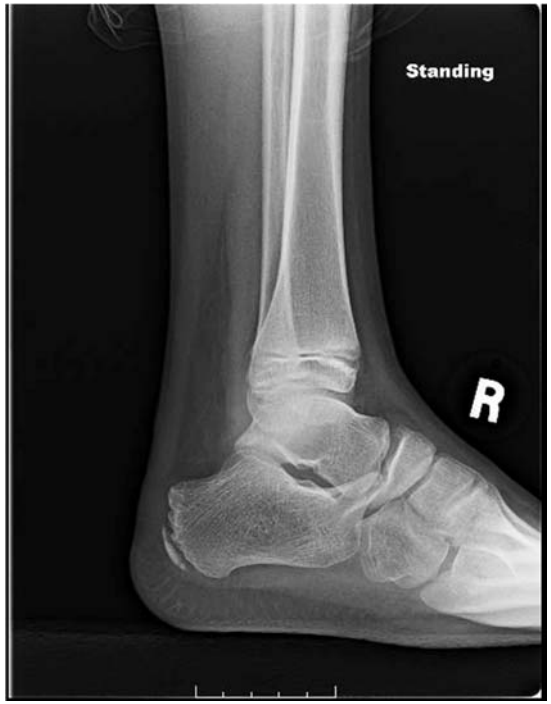
Figure 1:

Fig 1A: 

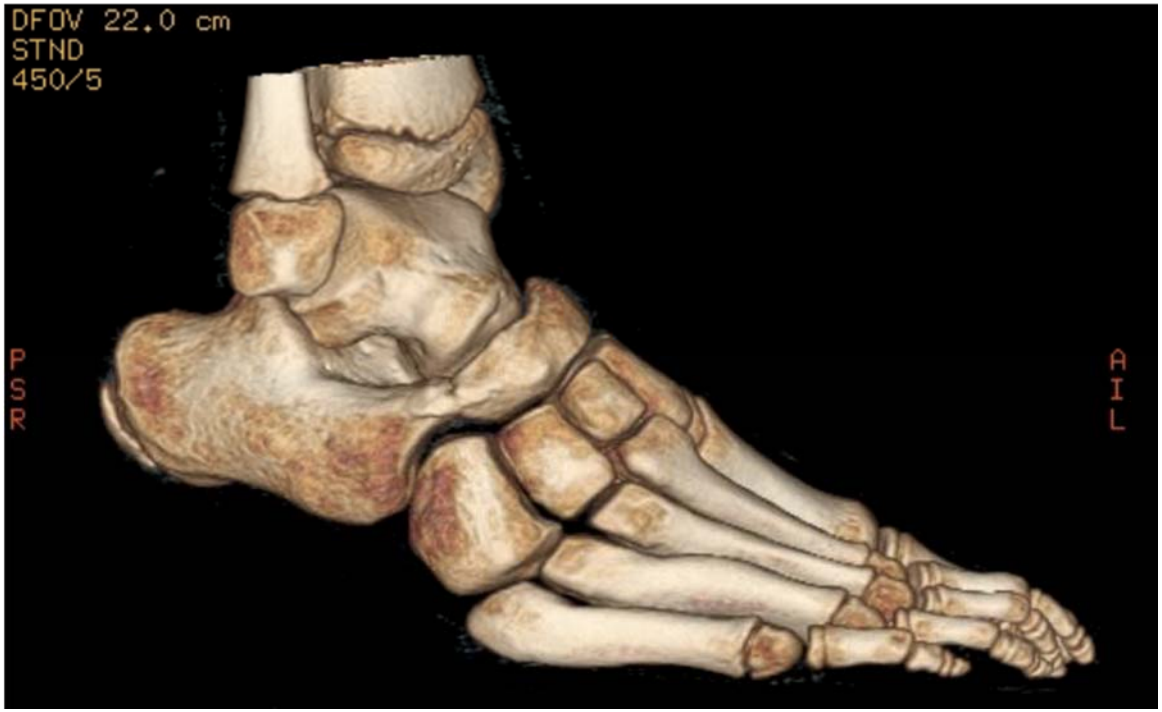


Fig. 1B:

Figure 2:

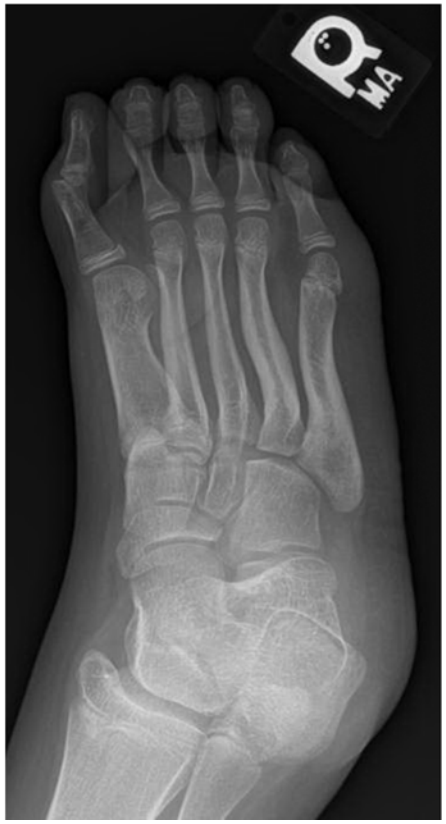
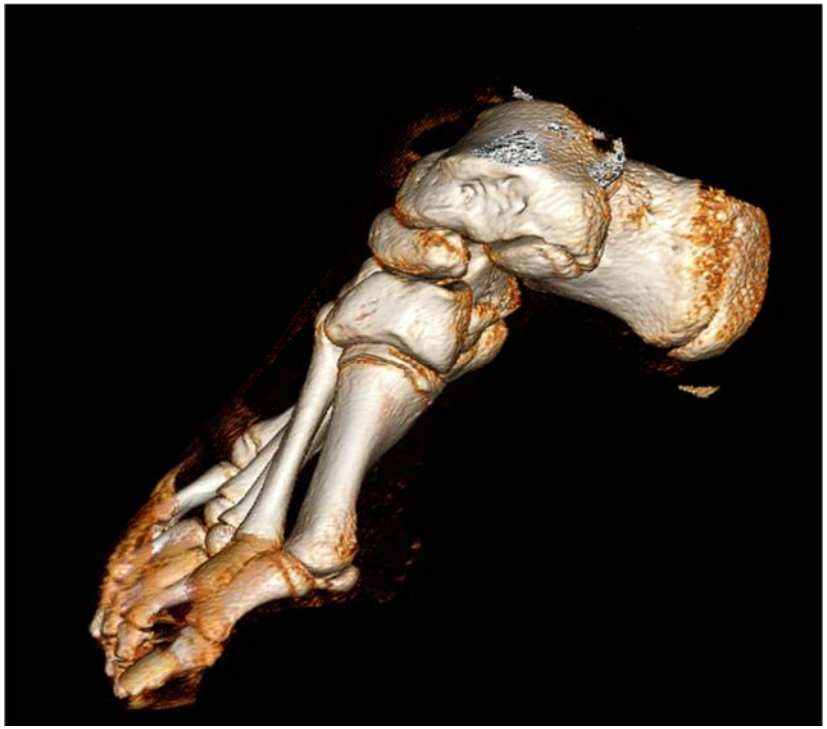
Fig 2A: 

Fig 2B: 

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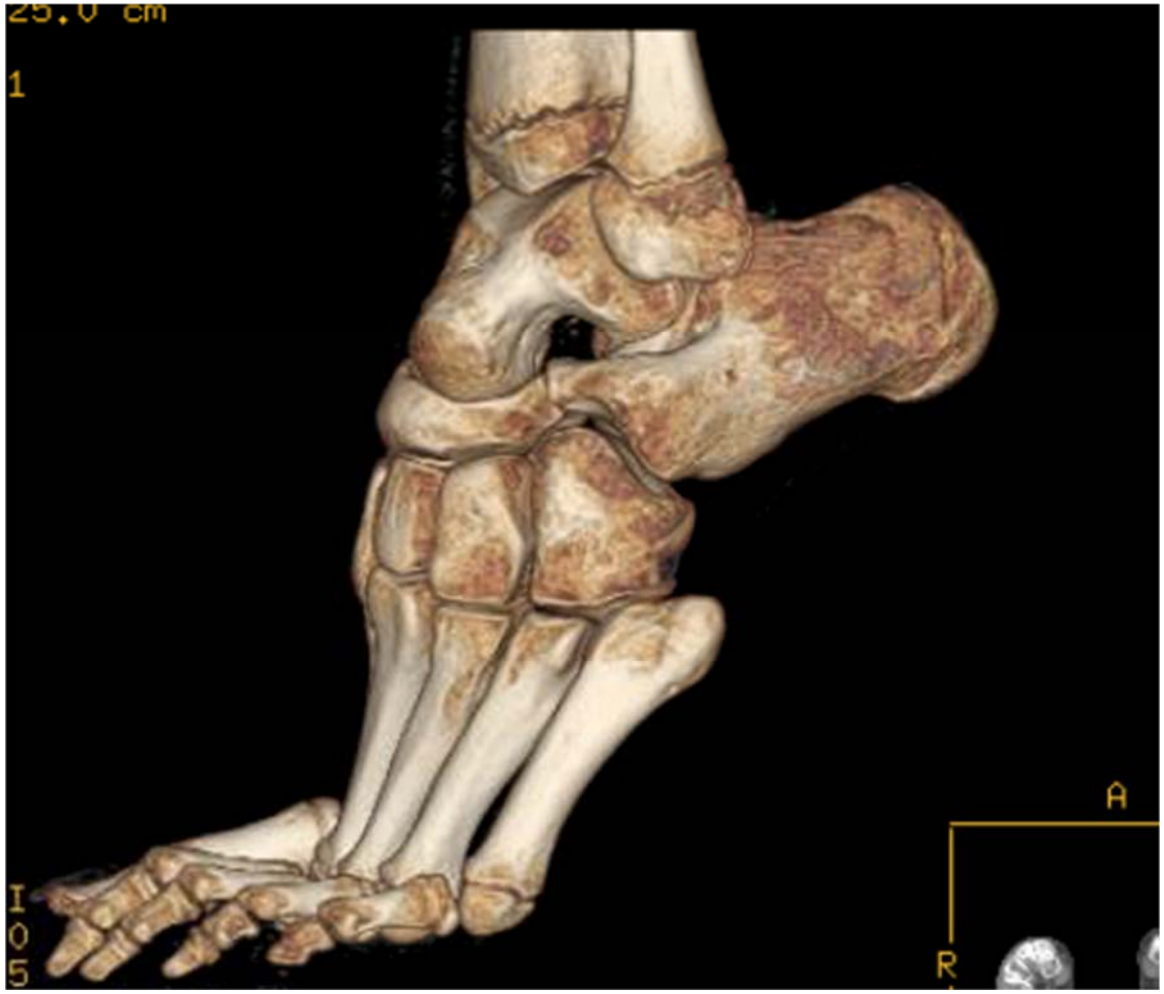

Fig 3:

Figure 4:

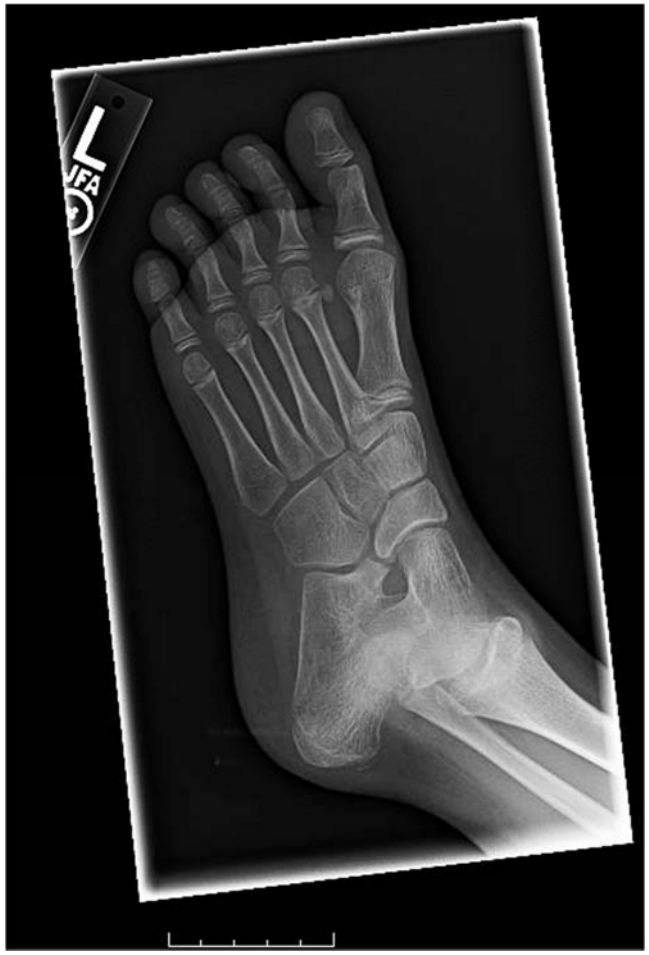

Fig 4A:

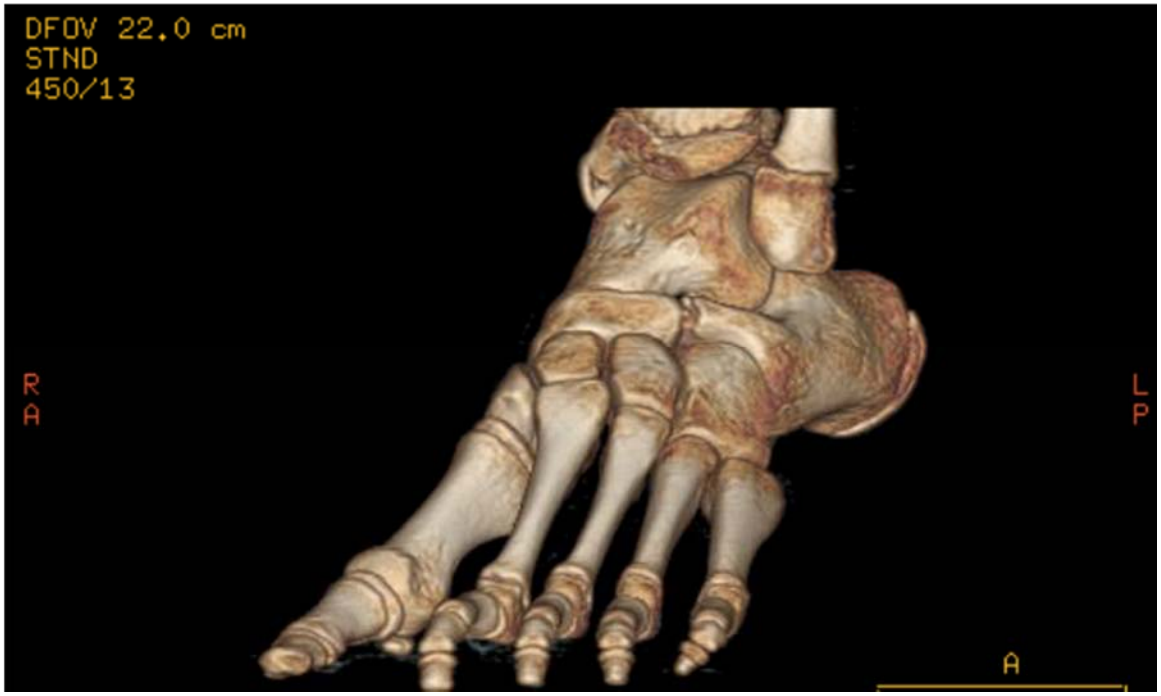

Fig 4B: